Case Report

Surgical management of chronic sixth cranial nerve palsy: case report and literature review

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ABSTRACT

Background: Esotropia resulting from sixth cranial nerve palsy can substantially impact an individual's visual acuity and overall quality of life. If the condition does not resolve in 6–10 months, surgical intervention may be necessary. Various muscle surgeries may be considered, with vertical rectus muscle transposition emerging as the primary option for treatment of complete palsy. However, this technique carries the risk of anterior segment ischemia and post-surgery deviations. Herein, we present a successful treatment of chronic complete sixth nerve palsy using a modified Nishida procedure, without splitting or tenotomy, and an adjunct botulinum toxin A (BTA) injection in the ipsilateral medial rectus muscle.

Case Presentation: A 59-year-old woman with a history of traumatic sixth nerve palsy had previously undergone horizontal muscle strabismus surgeries. Following multiple left medial rectus recessions, lateral rectus resection, and BTA injections, esotropia persisted. The worsening of her condition led to emotional distress and impaired social interaction. Initial examination revealed marked esotropia and limited left eye abduction. Magnetic resonance imaging (SIGNA MR750w, GE Healthcare, Waukesha, WI, USA) of the left eye revealed a contracted medial rectus muscle and substantial atrophy of the left lateral rectus muscle. A modified Nishida procedure was performed with an injection of 3 units of BTA into the ipsilateral medial rectus muscle, resulting in improved ocular alignment and stable findings after nine postoperative months. Furthermore, we supported our successful outcome with a summary of similar reported cases of sixth nerve palsy managed using the modified Nishida procedure with or without adjunctive procedures.

Conclusions: Following the modified Nishida procedure, the patient experienced a reduction in diplopia, improved ocular alignment and stability, and an increased binocular diplopia-free field. This case underscores the importance of an individualized approach to complex strabismus cases and highlights the modified Nishida procedure as a valuable tool in such circumstances. In the future, strabismus management will focus on refining personalized treatment and exploring innovative techniques for complex cases. Our success in using a combination of Nishida procedure and BTA injection should be further investigated in large-scale studies.

KEYWORDS

secondary esotropia, traumatic sixth-nerve palsy, noncomitant strabismus, horizontal diplopia, ocular surgery, modified nishida procedure, botulinum toxin, botox, botulinum neurotoxin type a

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INTRODUCTION

Esotropia secondary to sixth cranial nerve palsy, characterized by severe inward deviation of the eye, substantially impacts an individual's visual function and quality of life [1]. When a patient presents with a long-standing complete sixth nerve palsy, the treatment options are limited. In many cases, surgical intervention is required to correct the eye misalignment and improve visual function [2, 3]. If the palsy does not resolve within 6–10 months, surgical intervention is frequently required [2-5]. Choosing the best surgical strategy requires assessing the function of the lateral rectus (LR) muscle and angle of deviation. Nevertheless, LR function assessment may be impacted by medial rectus (MR) muscle contracture, resulting in an incorrect diagnosis of complete LR palsy [2]. Other tests that can distinguish between partial and complete LR palsies include the forced duction test, force generation test, saccadic velocity, botulinum toxin A (BTA) injection, and evaluation of the post-injection abduction facility [6, 7].

Surgery on the horizontal muscles is generally considered in cases with some preserved LR function. Options for this type of surgery include contralateral MR recession for significant incomitance, ipsilateral LR resection, and ipsilateral MR recession [8, 9]. In contrast, complete LR palsy necessitates a different strategy, with the main surgical objectives being the expansion of the binocular diplopia-free field, improvement of the abduction deficit, and eye alignment in primary position [4, 6]. Complete sixth nerve palsy is now primarily treated with vertical rectus muscle transposition (VRT); the two most common VRT techniques are those of Jensen and Hummelsheim [2]. The procedures have equal effects and complication rates in managing esotropia due to chronic sixth nerve palsy [10].

Anterior segment ischemia (ASI), a risk associated with VRT, may cause vertical and tortional deviations post-surgery [8, 9, 11]. Johnston and Crouch first suggested the potential advantages of transposing only the superior rectus muscle, with or without MR recession, to minimize the extent of surgery and theoretical risk of ASI. This approach has gained popularity recently, with Hunter further incorporating MR recession with an adjustable suture [7]. Many other modifications can be considered in VRT, including using scleral augmentation sutures or muscle union sutures, resection of a portion of the transposed muscle, and a modified Nishida procedure (no split, no tenotomy transposition) [2-4].

We report the successful management of chronic complete sixth nerve palsy using a modified Nishida procedure, without splitting or tenotomy, and an adjunct BTA injection in the ipsilateral MR muscle.

CASE PRESENTATION

A 59-year-old woman presented with long-standing left eye esotropia and face turning resulting from head trauma sustained at 24-years-old. The patient also reported experiencing troublesome horizontal diplopia, which further affected her daily functioning and emotional well-being. She had undergone horizontal muscle surgeries for sixth nerve palsy in the years 1987 and 1999; however, the outcomes were unsatisfactory and the condition gradually deteriorated. The previous surgeries included a right MR recession of 6 mm with a right LR resection of 9 mm, and afterward, a right LR re-resection of 4 mm. Despite multiple surgeries performed on both horizontal muscles, a substantial under-correction remained.

Upon examination, the patient had a best-corrected distance visual acuity of 20/30 in the right eye and 20/40 in the left eye using a Snellen chart (Nidek auto chart projector CP 670; Nidek Co., Ltd., Gamagori, Japan). Detailed anterior and posterior segment examinations under a slit lamp (Photo-Slit Lamp BX 900; Haag-Streit, Koeniz, Switzerland) and intraocular pressure measurements (Goldmann applanation tonometer AT 900°, Haag-Streit) revealed normal findings in both eyes. A Hirschberg test [12] showed left eye esotropia measuring 90–100 prism diopters (PD) at distance and near on a modified Krimsky test [13] with abduction limitation of -6 in the left eye and adduction deficit in the right eye in levoversion (Figure 1). The forced duction test [14] result was positive for the left MR muscle. The remaining cranial nerve examinations revealed normal findings.

Magnetic resonance imaging (SIGNA MR750w, GE Healthcare, Waukesha, WI, USA) [15] of the left eye revealed a contracted MR muscle and substantial atrophy of the LR muscle (Figure 2).



Figure 1. Preoperative ocular alignment photographs revealing left eye esotropia in primary position measuring 90–100 prism diopters prior to modified Nishida surgery. The images show limited abduction of - 6 in the left eye secondary to long-standing left sixth nerve palsy along with adduction deficit in the right eye in levoversion.



Figure 2. Axial T1-weighted magnetic resonance imaging (SIGNA MR750w, GE Healthcare, Waukesha, WI, USA) of the brain and orbits without fat-suppressed contrast enhancement. The image shows left eye esotropia with contracted medial rectus muscle and fibrotic and atrophied lateral rectus muscle (yellow arrow).

Considering the complexity of the case, a modified Nishida procedure [4] using nonabsorbable sutures was selected as the surgical approach. A 3-unit injection of BTA (Botox*; Allergan, Parsippany, NJ, USA) [16] (Figure 3A) was administered into the left (previously recessed) MR muscle 8 mm posterior to the insertion while the muscle was firmly grasped using conjunctival forceps [17]. According to the modified Nishida procedure [4], superior and inferior cul-de-sac conjunctival incisions were created to expose the rectus muscles. Ethibond 6-0 polyester sutures (Ethicon Inc., Somerville, NJ, USA) were attached to the temporal one-thirds of the superior and inferior recti 8 mm behind their insertions. Then, the sutures were attached 12 mm posterior to the superotemporal and inferotemporal quadrants of the limbus using a scleral bite (Figure 3A). Subsequently, the temporal margins of the muscles were sutured to the sclera in the superotemporal and inferotemporal quadrants 12 mm from the limbus, midway between the LR and vertical muscle insertions (Figure 3B). The aim was to improve ocular alignment and expand the binocular diplopia-free field to reduce the patient's troublesome horizontal diplopia.



Figure 3. The modified Nishida procedure (A and B), uses (A) Ethibond 6-0 polyester sutures (Ethicon Inc., Somerville, NJ, USA) to attach the temporal one-thirds of the superior rectus (SR) and inferior rectus (IR) muscles to the superotemporal and inferotemporal sclera, respectively, 12 mm behind the limbus, using a scleral bite [4] (A and B). As an adjunct (A), 3 units of botulinum toxin A (BT-A, Botox*; Allergan, Parsippany, NJ, USA) was injected into the left (previously recessed) medial rectus muscle 8 mm posterior to the insertion while firmly grasping the muscle with conjunctival forceps [16, 17].

Following the modified Nishida procedure, the patient achieved orthophoria in the primary position. No surgical complications were observed, and there were no symptoms of ASI [18]. The patient's examination findings remained stable after 9 months' follow-up, indicating success of the surgical intervention in correcting ocular malalignment and troublesome diplopia (Figure 4). Since levoversion in left eye abduction was not completely recovered, diplopia in levoversion persisted; however, with a slight head turn, the symptoms were tolerable.

This case report received ethical approval at the department level, and written informed consent was obtained from the patient for the publication of this case report and accompanying images.



Figure 4. Ocular alignment photographs at the 9-month postoperative examination. The patient has orthotropia in the primary position (near and distance) with mild improvement of abduction in levoversion. Despite the transportation process, vertical duction was well preserved. These photographs show the mild improvement of abduction in levoversion but continued limited abduction.

DISCUSSION

Using the modified Nishida procedure with an adjunctive BTA injection into the ipsilateral antagonist muscle, we successfully treated a patient with severe LR muscle atrophy and long-standing left eye esotropia due to traumatic sixth nerve palsy.

The management of paralytic strabismus presents a challenge for ophthalmologists, particularly in complex cases such as long-standing esotropia with severe muscle atrophy [19, 20]. For complete sixth nerve palsy, currently, VRT procedures are the first-line treatment [2]. They can maintain the LR ciliary arteries and enhance binocular vision field and abducting force. Despite some variability in results, several authors, including Hummelsheim, O'Connor, Jensen, and Foster, have described superior and inferior rectus transpositions on multiple occasions with notable deviation improvements [2]. In 1997, Foster introduced a modification known as posterior fixation of transposed muscles [21]. When the temporal one-fourth width of the superior and inferior rectus bellies are fixed to the upper and lower one-fourth widths of the LR belly, respectively, using 6-0 nonabsorbable sutures at 7–8 mm posterior to the LR insertion, this is known as an intermuscular augmentation suture, or Wright modification [2]. New vertical and torsional deviation and ASI are possible side effects of these procedures [2-4]. Following the modified Nishida procedure, our patient achieved orthophoria in the primary position. No surgical complications were observed, and there were no symptoms of ASI

The modified Nishida procedure has become the primary in treating full LR palsy [2, 4]. The objectives of this procedure are to lessen diplopia, increase the field of binocular vision, and enhance ocular alignment [2]. Furthermore, it is used to manage type 1 Duane syndrome [22] and MR hypoplasia [23]. For sixth nerve palsy, Muraki et al. [8] created a novel transposition method that eliminates the need for muscle splitting or tenotomy. To the best of our knowledge, our case represents the first successful treatment of long-standing sixth cranial nerve palsy—and longest-standing documented case—using a modified Nishida procedure and BTA injection into a previously recessed MR muscle after multiple previous horizontal muscle surgeries. In a case report by Mata Moret et al. [16], a 39-year-old man with long-standing bilateral sixth cranial nerve palsy secondary to a severe head injury 15 years prior was successfully treated using the Nishida procedure combined with a 10-unit BTA injection into the MR [16]. However, we conducted the procedure on a patient with a much longer, 35-year history of traumatic sixth cranial nerve palsy and multiple previous surgeries that resulted in fibrotic and tight muscles. The case reported by Mata Moret et al. [16] had no previous ocular surgery.

Sixth nerve palsy can be treated using the modified Nishida procedure, which does not require splitting or tenotomy during transposition [8]. However, recent research suggested that the procedure can also be used for causes of vertical strabismus, including inferior rectus and inferior oblique aplasia; monocular elevation deficiency; and transection, or disinsertion of the inferior rectus muscle [2, 11]. This procedure lowers ASI risk and is shorter and simpler than conventional VRT. Our literature review revealed only one reported ASI case [24]. Our patient had no postoperative complications, including ASI.

Hernandez-Garcia included 32 out of 43 patients who had sixth nerve palsy [6]. The modified Nishida technique alone corrected a mean (standard deviation) of 29.4 (6.6) PD, and adding an MR muscle recession corrected 62.6 (23.8) PD. MR recession, BTA, and the modified Nishida procedure corrected 95.0 (18.0) PD overall. Depending on passive duction and preoperative horizontal deviation, it is possible to determine the necessity of additional procedures, such as MR muscle recession and BTA, which increase the effect in primary position [6]. Likewise, the series of patients with complete LR palsy reported by Murthy et al. had good outcomes using this technique [25]. The adjunct procedure in the current case was a BTA injection into the ipsilateral MR muscle.

The modified Nishida procedure yielded positive results in our case. The patient's binocular vision improved and orthophoria in the primary position was attained. Follow-up visits revealed no surgical complications, and the results remained stable up to the final visit at 9 months. This suggests effectiveness and safety of the modified Nishida procedure in treating complicated cases of chronic paralytic strabismus. The procedure improves ocular alignment and restores force balance by transposing the superior and inferior rectus muscles [26]. This method reduces the need for repeat procedures and yields durable results; according to previous reports, there are sixth nerve palsy cases with 9–20 months' evolution from onset [4, 8, 16, 25] (Table 1). The present case represented a long-term, 35-year palsy in previously operated horizontal muscles with significant scars surrounding the MR and LR muscles and a large angle. Nevertheless, the results were comparable to those reported in other publications [3, 4, 6, 8, 16, 24, 25, 27] (Table 1). Furthermore, the current case responded well to BTA even though it is not recommended in palsy cases that have progressed beyond 6 months [28].

Author (Year)	Patient's age and sex	Mean preoper- ative deviation (PD)	Mean duration of deviation (m)	Adjunctive procedure	Follow-up duration (m)	Final outcome
Current study	A 59-year-old woman.	90–100	420	MR botulinum toxin injection.	9	Orthophoria in primary position.
Hernandez-Garcia et al. (2023) [6]	Thirty-two patients (18 men and 14 women). The mean \pm SD (range) age was 38.6 \pm 19.8 (3 to 82) years.	63.0 ± 27.3 (range: 24–125)	-	MR recession with or with- out botulinum toxin injection.	6	The mean ± SD (range) in primary position was 2.8 ± 9.2 (- 30 to 20) PD.
Moledina et al. (2023) [24]	A 68-year-old wom- an with bilateral sixth nerve palsy.	35	7	-	6	Residual right esotropia in primary position.
Mata Moret et al. (2019) [16]	A 39 year-old man with bilateral sixth nerve palsy.	45	180	MR botulinum toxin injection.	12	Orthotropia in primary position.
Sabermoghadam et al. (2018) [3]	Ten patients (6 men and 4 women). The mean ± SD (range) age was 44.2 ± 9.2 (28 to 57) years.	The mean ± SD (range) was 49.5 ± 9 (40–65).	8–16	MR recession.	6-12	Orthotropia to 12 PD of esotropia.
Murthy (2017) [25]	A 7-year-old girl.	90–100	-	MR recession.	1	Exophoria in prima- ry position.
	A 31-year-old man.	35 for distance 25–30 for near	9	-	6	Orthotropia.
	A 43-year-old woman.	50	12	MR recession.	3	Orthotropia.
Sharma et al. (2017) [27]	Twenty-one patients with nonresolving sixth nerve palsy.	The mean \pm SD was 57.64 \pm 24.36 in nonadjustable and 56.0 \pm 23.66 in adjustable MR recession group.	-	Nonadjustable or adjustable MR recession.	6	Esophoria.
Muraki et al. (2013) [8]	Nine patients (5 men and 4 women). The mean ± SD (range) age was 53.8 ± 9.6 (31 to 66) years.	The mean ± SD was 52.2 ± 21.0.	The mean \pm SD (range) was 36.0 \pm 47.5 (12 to 151), except for 1 patient who had left sixth nerve palsy at birth.	MR recession in 6 of the 9 patients.	The mean ± SD (range) was 14.3 ± 28.3 (1 to 89).	The mean ± SD ocular deviation was + 5.9 ± 12.9 PD.
Nishida et al. (2005) [4]	A 66-year-old woman.	75	-	MR recession.	-	Residual esotropia of 25 PD in right gaze.

$Table \ 1. \ Studies \ on \ the \ modified \ Nishida \ procedure \ in \ management \ of \ sixth \ nerve \ palsy \ with \ or \ without \ adjunctive \ procedures$

Abbreviations: PD, prism diopter; m, months; MR, medial rectus; SD, standard deviation.

We have presented our case of long-standing paralytic esotropia managed using a modified Nishida procedure with a single BTA injection into the ipsilateral antagonist muscle. This study has limitations. Its restriction to a single case and the lack of strong evidence proving the efficacy of our treatment. However, we supported our successful outcome with a summary of similar reported cases of sixth nerve palsy managed using the modified Nishida procedure with or without adjunctive procedures. Using BTA is not recommended in chronic sixth cranial nerve palsy management; therefore, we could not determine with certainty the effectiveness of this adjunctive modality. Larger, well-designed studies with detailed preoperative clinical and paraclinical evaluations and longer follow-up periods may provide robust insight into the applicability of BTA in this clinical setting. These studies could provide the optimal management approach for improving the binocular field and resolving troublesome diplopia in challenging cases of chronic sixth cranial nerve palsy.

CONCLUSIONS

This case study describes the effective management of a challenging strabismus case with severe LR muscle atrophy and long-standing left eye paralytic esotropia with previous horizontal muscle surgeries. The patient experienced less diplopia after the modified Nishida procedure with BTA injection into the MR, with improved ocular alignment and stability and an increased binocular diplopia-free field. This highlights the value of an individualistic approach in complex cases of strabismus and emphasizes the modified Nishida procedure as a useful tool in these circumstances. Our success in using a combination of Nishida procedure and BTA injection should be further investigated in large-scale studies.

ETHICAL DECLARATIONS

Ethical approval: This case report received ethical approval at the department level, and written informed consent was obtained from the patient for the publication of this case report and accompanying images. **Conflict on interests:** None.

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